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Stakeholder views about participating in paediatric biobanks: a narrative review

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1Abstract

2Scientific and medical research with children is essential to the
3development of therapies for younger patients. Paediatric biobanking uses
4samples from minors to provide a critical and expanding resource for such
5research. It also raises important ethical, legal and social implications
6(ELSI) and highlights the need for appropriate frameworks for practice
7developed through stakeholder consultation. This paper reviews the
8current literature on stakeholder views of paediatric biobanking. A
9narrative review was conducted of empirical studies in this area, the
10majority of which did not involve actual biobanks or include the views of
11children. Key themes were identified: parental consent; childrens' assent;
12the return of results; and risks, benefits and burdens of participation. The
13resultant analysis highlights how children involved in paediatric biobanks
14are not only capable of understanding and assenting to their participation
15but also of contributing their voices to the development of future
16paediatric biobanking policy.

17**Key words:** paediatric biobank, children, ethics, narrative review, genetic,
18social

11. Introduction

2The use of human samples in biomedical research has resulted in
3unprecedented scientific breakthroughs (Wolf, Bouley et al. 2010).
4Biobanking technology today offers a rich resource for studying many
5aspects of human health and disease. The Organisation for Economic Co-
6operation and Development defines biobanks as “a collection of biological
7material and the associated data and information stored in an organised
8system, for a population or a large subset of a population” (Sampogna
92006). Paediatric biobanking promises greater understanding of the
10causes, prevention and treatment of childhood disease (Brisson, Matsui et
11al. 2012). The ongoing dearth of knowledge of preventive measures and
12appropriate treatment for children at different stages of their development
13is internationally recognised (Gill 2004). This limitation results in part from
14stringent child protection restrictions placed on research involving children
15(Avard, Silverstein et al. 2009). But alongside the need for child protection
16in research is an equal and growing need for the inclusion of child
17participants in order to generate age-specific findings (Field and Behrman
182004). Paediatric biobanks continue to be developed as a basis for
19facilitating medical and scientific research with samples from child
20populations (Samuël, Knoppers et al. 2012). Their development has been
21accompanied by a corresponding interest in the ethics of children’s
22participation in research and a need for sociological inquiry into practices
23connected to it (Avard, Sénécal et al. 2011). Future understandings can be

1expected to aid in policy development and the implementation of
2culturally appropriate paediatric biobanking practices.

3Paediatric biobanking is governed by myriad restrictions and regulations,
4an unsurprising situation given the historical injustices in bioscience
5perpetrated on vulnerable populations (Avard, Silverstein et al. 2009) and
6the controversial discovery of unconsented bio-repositories such as those
7at Alder Hey hospital and Bristol Royal Infirmary in the UK (Boden, Epstein
8et al. 2009, Avard, Sénécal et al. 2011). There is also ongoing limited
9guidance on promoting good biobanking practices involving samples from
10children (Samuël, Knoppers et al. 2012). In paediatric biobanking,
11participants are in a state of growing maturity requiring researchers to
12address issues of privacy, autonomy and concepts of risk that all change
13with age. Some argue that research on children should only be undertaken
14if it involves no more than minimal risks, namely those no higher than
15what a child would encounter in daily life (Hens, Nys et al. 2009). A
16dynamic approach to paediatric biobanking has also been put forward
17(Avard, Silverstein et al. 2009), one founded on continued communication
18among all stakeholders (for example, the model proposed by (Kaye,
19Whitley et al. 2014)).

20Children are key stakeholders and have demonstrable knowledge of the
21benefits and challenges of biobank participation (Anderson, Stackhouse et
22al. 2011). Considering their views in research recognizes their worth as
23human beings (Roberts 2008) and potentially improves the protection of
24children while fostering trust in biobanking (Lemke, Wolf et al. 2010). Such

1consultation may also help highlight pitfalls in research design and
2provide opportunities for improving research (Godard, Marshall et al.
32004). The *UN Convention on the Rights of the Child* states that children
4capable of forming their own views should be assured their right to
5express these views on matters concerning them (Morrow and Richards
61996). The *Children's Act of 1989* in England and Wales stipulates that
7courts shall regard the wishes and feelings of the child, while professional
8bodies such as the British Psychological Association and the British
9Sociological Association as well as research organisations such as the
10Medical Research Council encourage active consultation and engagement
11with children involved in research.

12Little is known, however, about children's understandings of their social
13positioning within health research (Mayall 1998). Children are increasingly
14seen as competent social actors worthy of study in their own right (James
15and James 2001), and that understanding childhood requires revealing the
16experiences of children (Shanahan 2007). But giving voice to children
17entails more than simply letting children speak; it necessitates a deeper
18exploration of their experiences and how adults theorise these (James
192007). Understanding social order from a child's standpoint requires
20studying not only a child's perceptions but also the development of the
21concepts that underpin such knowledge (Mayall 1998). Social studies of
22paediatric biobanking is therefore needed to inform more fully policies
23affecting the study of health and disease in children (Grover 2004).

1While paediatric research must always be finely balanced against child
2protection (Mumford 1999), overweighting in favor of protectionism can
3result in ‘therapeutic orphans’ and a limited number therapies tested for
4safety and efficacy in children (Samuël, Ries et al. 2008). The construction
5of children’s vulnerability may also lead some researchers to view child
6participants as ‘out-of-bounds’ with the result that their voices are
7silenced (Moran-Ellis 2010). Past efforts to advance research while
8maintaining effective child protection have rested on the principle of
9subsidiarity; that is, permitting research on children only if it cannot be
10done on adults (Hens, Van El et al. 2012). Research with children has also
11generally been based on the precautionary principle that adequate
12measures must be taken to avoid potentially harmful outcomes when
13there is an expectation based on empirical evidence or causal hypotheses
14that damage could occur (Jarosinska and Gee 2007). This approach
15demands full consideration be given to any physical and emotional harm
16to the child, such as avoiding venipuncture by using residual blood from
17diagnostic testing, or conducting data collection at home or a familiar
18setting rather than in a hospital (Avard, Silverstein et al. 2009). Risks
19specifically associated with biobank participation include breaches of
20privacy, the disclosure of information to third parties, and possible
21stigmatisation of participants based on genetic results (Avard, Silverstein
22et al. 2009).

23The objective of this paper is, therefore, to identify and critically review
24existing empirical research into the views and perspectives of principal
25stakeholders involved in paediatric biobanking – namely children, parents

1and researchers. The authors aim to summarise not only key findings
2emerging from this literature, but also to critically examine higher level
3ethical, legal and social implications (ELSI) issues cross cutting the
4existing evidence base. The authors assume from the outset that
5children's voices are essential to future development of paediatric
6biobanking policies and best practice.

72. Methods

8A narrative approach was used to review the literature. The aim was to
9identify empirical research involving stakeholders and/or participants in
10paediatric biobanks. A narrative rather than systematic review was
11undertaken for reasons outlined in the literature (Petticrew and Roberts
122008, Bryman 2012), namely: that the focus of the review was broad
13rather than specific; the studies under consideration were largely
14qualitative or mixed methods; and the objective was to assess individual
15studies rather conduct a meta-analysis. A literature search was conducted
16by the lead author (CO) to identify all articles published in English prior to
17May 2014 using multiple bibliographic databases. The search process was
18iterative and continued until no new articles were found (Petticrew and
19Roberts 2008). The overall strategy was additionally reviewed for quality
20and output by a second author (JM), who is a professional librarian.

21In order to pinpoint search terms most applicable in the field of paediatric
22biobanking, an initial pilot search was conducted using the Web of
23Knowledge (WoK) bibliographic database. An analysis of the results from

1exploratory searching determined that virtually all relevant papers
2included keywords on two themes: children and genetic databases. There
3was, however, considerable variation in terminology use. The terms
4relating to children were *child/children, minor, youth, young people,*
5*adolescent* and *paediatric*; while those relating to genetic databases were
6*biobank(s), gene bank(s), gene repository/ies, genetic database(s), stored*
7*DNA* and *genomic database(s)*. Based on these pilot efforts, an initial
8search was conducted in WoK using the terms identified above for children
9and genetic databases. The final search used was:

10TOPIC: (child* OR minor OR youth OR young people OR adolescent OR
11paediatric) AND

12TOPIC: (biobank* OR gene bank* OR gene repositor* OR genetic
13database* OR stored DNA OR genetic repository* OR genomic database*)

14The search (along with all subsequent updating searches) was conducted
15without date restrictions because paediatric biobanking remains a
16relatively new practice whose documented evidence base is small. Initially
17we focused on general population biobanks that either included mainly or
18only children. As these efforts yielded few papers, it was decided that
19disease-specific tissue banks should also be included to elicit a range of
20views about children's participation in biobanking more broadly.

21The final WoK search (May 2014) produced 311 unique hits. These were
22assessed by reviewing each paper's keywords and abstract using
23predefined inclusion and exclusion criteria. Papers were included for

review if they reported findings from empirical studies into people's opinions, views, perceptions or experiences with a paediatric biobank, paediatric tissue bank, or any biobank and tissue bank including or intended to include samples from children. Papers excluded from review were those of a medical laboratory nature, those detailing the structures of biobanks, any papers based on blood spots or Guthrie cards, those focusing on predictive genetic screening, and publications not based on empirical studies (e.g. theoretical papers, systematic reviews, legal document reviews). Papers were also excluded from review if they were not published in English or if full-text versions could not be obtained. Some papers included for review sought participants' views on the idea of paediatric biobanking rather than actual paediatric biobanks. For the purposes of this paper these were categorised as being "hypothetical" biobanks owing to the fact that the biobanks either did not exist or were being established but not yet operational.

The WoK search was then repeated in PubMed (119 hits) and Scopus (195 hits). A total of 11 articles met the inclusion criteria. Following all three searches, a similar and final search was conducted using Google Scholar (GS). Because GS offers a less precise search interface, the search strategy incorporated additional terms relating to research methods and study type. Of the terms used, the first was 'paediatric biobank', the second focused on research methodology (e.g. qualitative, empirical, thoughts, views, experiences) and the third related to study type (e.g. birth cohort, longitudinal). Within GS the final search used was:

1 *paediatric biobank AND qualitative OR empirical OR thoughts OR views*
2 *OR experiences AND longitudinal birth cohort*

3 Two further articles were identified using GS. To ensure a comprehensive
4 literature review, a 'pearl growing' (Bryman 2012) assessment of the
5 references cited in all 13 articles was carried out. This produced two
6 further articles that met the inclusion criteria, for a total of 15.

7 When conducting a narrative review, Bryman (Bryman 2012) also
8 recommends reviewing key authors' publications. Based on papers
9 assessed for inclusion, several researchers were identified as prominent in
10 the broader field of paediatric biobanking through their involvement in
11 either empirical or theoretical research. A search for each was conducted
12 in WoK and PubMed, identifying one further paper. Corresponding authors
13 of all 16 papers were contacted by CO to inquire if further work had been
14 completed or published. This yielded six papers for a total of 22. Emails
15 were also sent to 70 paediatric biobanks and biobanks involving families
16 requesting references, though no new articles meeting the inclusion
17 criteria were identified.

18 **3. Results**

19 The results section first provides a brief overview of the papers and the
20 different types of research undertaken. Secondly we identify four main
21 themes emerging from the findings and considers their implication for
22 future research and practice in paediatric biobanking.

23 **3.1. Summary of papers reviewed**

1All papers reviewed are summarised in Table 1. Four key characteristics
2were identified among them. First, the 22 papers reported only 17
3empirical studies (papers reporting on single underlying studies were:
4Study A (Halverson and Ross 2012, Lemke, Halverson et al. 2012); Study B
5(Goodenough, Williamson et al. 2003, Goodenough, Williamson et al.
62004, Williamson, Goodenough et al. 2004); and Study C (Hens and
7Dierickx 2010, Hens, Nys et al. 2010). Second, only one study sought
8solely children's views about their experience of participating in biobank
9research (Goodenough, Williamson et al. 2003, Goodenough, Williamson
10et al. 2004). Two others (Dixon-Woods, Wilson et al. 2008, Harris, Ziniel et
11al. 2012) included both children and adults, though these were based on
12tissue bank research (i.e. disease specific biobanks); in these papers the
13contribution of children was not made explicit in the findings. One study
14included adolescents (i.e. children in their teens) as well as adults (Hens
15and Dierickx 2010, Hens, Nys et al. 2010). Third, a clear majority of the
16papers (n=13) involved what we have termed hypothetical paediatric
17biobanks (i.e. biobanks that did not exist at the time of the study). Finally,
18almost all authors referred to children, minors or adolescents without
19specifying age ranges (for this paper, we use *child* or *children* to refer to
20any individual under 18 unless specified otherwise by an author).

21 3.2. Themes arising from the review

22Four main themes emerged from the literature reviewed on paediatric
23biobanks. These were: (1) parental consent; (2) children's assent; (3)
24return of genetic results; and (4) risks, burdens and benefits of

1 participation. Each is discussed in turn, after which we briefly compare the
2 perspectives of the range of stakeholders involved in the reviewed
3 studies.

4 3.2.1. **Parental consent**

5 Parental consent was a recurrent theme in many of the papers. Although
6 never defined explicitly, parental consent was understood as parents'
7 agreement that samples, medical records and other information about
8 their children would come to be included in biobanks as well as agreement
9 to their child's ongoing participation. Consent was portrayed as a legally
10 binding agreement given by a parent or a child who had reached the age
11 of majority (usually age 16 years). Overall there was a general preference
12 for involvement in biobanks that used broad consent and simple consent
13 forms (Lemke, Halverson et al. 2012). While seeking views of women
14 about a hypothetical paediatric biobank Neidich et. al. (2008) found that
15 women supported the use of samples for a wide array of paediatric
16 conditions, either to help their own children or medical science more
17 generally. There was some variability regarding temporal restrictions on
18 broad consent, with some studies suggesting parents preferred re-contact
19 about future research (Lemke, Halverson et al. 2012), while others
20 indicated a more general willingness to sanction future research without
21 being re-contacted (McMurter, Parker et al. 2011).

22 Contrary to common practice in consent taking, parents reported a
23 preference for more straightforward and uncomplicated consenting
24 procedures particularly in the nature of the consent forms (Hens and

1Dierickx 2010). For example, a study proposing a hypothetical biobank
2(McMurter, Parker et al. 2011) found that parents of paediatric oncology
3patients would be satisfied to give a simple 'yes/no' consent to future
4research without the need for complicated consent forms. Another (Hens
5and Dierickx 2010) found researchers and healthcare professionals in
6genetic research believed that parents would have less confidence in
7research and be less likely to participate in paediatric biobanks if
8presented with long consent forms, suggesting more complex consent
9forms could possibly deter participation among parents.

10Parental consent on behalf of children was seen as potentially
11problematic. Professionals in the field of genetics felt that proxy consent
12(consent given by parents on behalf of children) could infringe on the
13child's rights by limiting the scope of the child's assent (Hens, Snoeck et
14al. 2010). Williamson et. al. (2004) postulated that power is negotiated
15between parents and children in a complex manner and that the position
16of children changes as they age and acquire more information.

17 3.2.2. **Children's assent**

18Children's assent was seen as the inclusion of a child's permission – or
19more simply, a child's agreement – to participate in research, once
20parental consent had been given on behalf of the child. All papers
21reported that children's views needed to be taken into account as part of
22paediatric biobanking practices. In particular, this theme identified a
23debate over how assent was managed empirically. The issues identified
24included: when to presume consent (age versus maturity) (Hens and

1Dierickx 2010); how to assent (Jackson, Dixon-Woods et al. 2009); whether
2to re-contact children to update consent (Goldenberg, Hull et al. 2009);
3and whether to make provisions for withdrawal from research (Ries,
4LeGrandeur et al. 2010). In their study involving parents of paediatric
5oncology patients, McMurter et. al. (2011) found that parents considered
6children capable of consent before the age of 18. Jackson et. al. (2009)
7affirmed this finding in their study of healthcare professionals but
8additionally found that strict formal adherence to assessing a child's
9capacity (such as requiring his/her signature or having a particular age set
10for majority) could interfere with the assessment of the child's ability to
11consent. Hens et. al. (2010) corroborated this finding in their study of
12professionals involved in paediatric biobanking, although their main
13departure from Jackson was their suggestion that 16 to 18 is a suitable
14age for children to consent. Jackson et al (2009) argued that specific age
15boundaries for consenting were too restrictive. Hens et al (2010) did,
16however, acknowledged the impracticality of assigning a fixed threshold
17for consent given the influence of an individual child's maturity and social
18context. Williamson et. al. (2004) reported that child participants viewed
19consent as a progressive relationship between themselves, their parents
20and researchers. Hens and Dierickx (2010) postulated that consent forms
21as used today mainly serve as a document for avoiding prosecution or
22litigation rather than being a document to improve patient-researcher
23relationship. Williamson et al. (2004) affirmed in their research with child
24participants of a biobank that children were cognisant of their changing
25priorities and opinions and could hence discuss their perceptions of their

own development logically. Additionally, in the case of long-term paediatric biobanking, competence and autonomy were thought to develop through direct social experience (Hens and Dierickx 2010).

Kaufman et. al. (2008) demonstrated that adults saw the importance of obtaining children's permission before inclusion in a biobank. Using a hypothetical scenario approach, Hens and Dierickx (2010) found that teenagers wanted to be re-contacted when participating in research for reasons of respect and curiosity rather than desire to control the research. The issue of re-contact for consent was also reported by Goldenberg et. al. (2009), who surveyed adult cancer patients about a hypothetical paediatric biobank that would include a sample from their childhood; in this case re-consent was seen primarily by respondents as indicative of the researchers' respect for participants' interest in research decision making. The debate on re-consent was presented as moving in tandem with a participant's ability to withdraw from a study (2008). While the right to withdraw upheld an individual's autonomy, it also restricted the potential for long term research. One reported way of avoiding high rates of withdrawal was to design participation in biobanks that allowed participants to waive future consent (Goldenberg, Hull et al. 2009). Williamson et. al. (2004) reported that some children felt pressure against dissenting to research participation in their study among child participants of a biobank.

3.2.3. Return of results to children or families

1Also identified in the analysis was a desire for the return of research
2results in paediatric biobanking (McMurter, Parker et al. 2011). There was
3variation in participants' expectations by way of returned results and how
4these would be communicated. Parents generally wanted some degree of
5feedback. For example, Harris et. al. (2012) found parents who had
6enrolled their children (with developmental disorders) in a tissue bank not
7only wanted to receive results but preferred to receive this information
8electronically, believing the results would help them understand their
9child's condition more fully. Additionally, Hens and Dierickx (2010) found
10that even though participants understood there to be a clear distinction
11between research and diagnosis, return of results was considered a
12humane act as compensation for research participation.

13Although parents generally wanted results returned, this desire led to
14secondary concerns, especially tensions between a child's autonomy and
15privacy vis-a-vis his/her parents (Hens and Dierickx 2010). The study
16found there was a need to strike a balance between a parent's desire for
17disclosure and a child's autonomy within research participation. This
18debate was also addressed by Hens et. al. (2010), who argued that
19communicating genetic results that lacked immediate medical value to
20parents would breach the principle of autonomy and affect the child's
21ability for self-governance. In addition to this, Harris et. al. (2012)
22postulated that a child's future autonomy in controlling their research
23results may be compromised when such results are disclosed to his/her
24legal guardian. Similarly, Hens and Dierickx (2010) argued that the rights
25of a child to know about (potentially treatable) medical conditions

1superseded the rights of his/her guardians in deciding not to know.
2Likewise, it was reported that returning genetic results could alter how
3parents treat their child (Hens, Nys et al. 2010). Such debates speak to
4ongoing concerns about best practices for upholding the autonomy of a
5child in paediatric biobanking.

6 3.2.4. **Risks, burdens and benefits of participation**

7The final theme addressed the balance between the risks and burdens of
8research and the potential benefits that could come from participation.
9While definition of terms such as *risks*, *burdens* and *benefits* was rarely
10made explicit in the papers, *risks* were mainly viewed as potential harms,
11*burdens* as excessive demands in time and effort, and *benefits* as possible
12gains from participation (including but not limited to financial or
13therapeutic gains). In the papers reviewed, the specific risks faced by
14children from biobanking were varied, from physical or emotional harm (a
15commonly reported example was venepuncture, though whether this
16meets the minimum risk threshold for child research participation may be
17debatable), to being uncomfortable socially with certain questions (e.g.
18questions about alcohol consumption among teenagers) (Hens and
19Dierickx 2010).

20A number of authors reported concern among researchers and the public
21to avoid risks and burdens for research participants. Others reported a
22similar finding among parents and children. Halverson and Ross (2012)
23specifically noted parents stating they would enrol their children in a
24biobank on condition of minimal risk to the children. Kaufman et. al.

1(2008) found parents in their study were not willing to enrol their children
2in biobanks because participation would be burdensome, especially if the
3biobank required daily recording of a child's life, vis a vis the already long
4list of activities of their children. Hens et. al. (2010) reported that the
5possibility of research being distressing or of limited benefit to
6participants could inhibit enrolment in biobanks. Public opinion, as
7presented by Kaufman et. al. (2008), revealed an expectation of benefits
8to individuals such as the child or the child's family, or a wider benefit to
9society through the advancement of medical knowledge. In spite of
10participants being concerned by the level of burden placed on them by
11participation, Lemke et. al. (2012) also found that parents believed the
12benefits of participating in a paediatric biobank outweighed the risks.
13Although parents wished to protect their children from unwarranted
14burden and risks, they were aware that certain risks are ubiquitous in
15society; and reasoned that no research is exempt from risk in the form of
16breaches (Lemke, Halverson et al. 2012).

17**4. Multiple stakeholders**

18A range of stakeholder opinions were covered in the papers included in
19this review. We note the specific patterns of these perspectives here. In
20papers reporting researchers' opinions, the issues resonating most were
21the need for both parental consent and child's assent (Jackson, Dixon-
22Woods et al. 2009, Hens and Dierickx 2010), the problematic nature of
23both blanket consent (Ries, LeGrandeur et al. 2010) and the return of
24research results (Ries, LeGrandeur et al. 2010), the need for privacy

1protection, and a concern that research should not be burdensome to the
2child (Hens, Snoeck et al. 2010). Papers reporting views of parents
3demonstrated that there was a general desire to receive research results
4(McMurter, Parker et al. 2011, Harris, Ziniel et al. 2012, Lemke, Halverson
5et al. 2012) and that children should be re-contacted as they matured
6(McMurter, Parker et al. 2011) for consent (Klima, Fitzgerald-Butt et al.
72013). Parents supported biobank use for a wide array of paediatric
8research (Neidich, Joseph et al. 2008) and their enrolment was based on
9their trust in biobanks (Neidich, Joseph et al. 2008, Brothers and Clayton
102012). Parents asserted that children should be given access to their
11health records (Halverson and Ross 2012). Papers reporting the views of
12the public had similar themes: the need for re-consenting after the child
13reaches majority (Kaufman, Geller et al. 2008, Goldenberg, Hull et al.
142009), the need for minimising a child's pain and burden in participation
15(Kaufman, Geller et al. 2008), a desire to receive research results
16(Halverson and Ross 2012), and the importance of trust in research
17participation (Halverson and Ross 2012). Papers involving both
18adolescents and adults (who were not participants in biobanks) reported
19similar findings, (though it is not clear which responses were from the
20adults and which ones were from the adolescents): that research should
21not be burdensome (Hens and Dierickx 2010); a trust in parents ability to
22consent to the right research as well as trust in biobanking (Hens, Nys et
23al. 2010); the importance of informed consent and growth toward
24autonomy (Hens and Dierickx 2010, Hens, Nys et al. 2010); the need for
25confidentiality; and, wanting meaningful research results communicated

1(Hens, Nys et al. 2010). Papers reporting the views of children alone or
2children and parents (actual biobank participants) expressed the need for
3both child's assent and parental consent (Goodenough, Williamson et al.
42004, Williamson, Goodenough et al. 2004) the need for anonymity
5(Goodenough, Williamson et al. 2004) and the growing autonomy of
6children (Goodenough, Williamson et al. 2004).

7**Discussion**

8This review highlighted four main areas of interest in paediatric
9biobanking. First, parents preferred simple consenting procedures, though
10it was noted that broad parental consent can be problematic if it impairs a
11child's future autonomy and control over their participation in research.
12Second, children's assent was considered important in research. The few
13children interviewed in these empirical studies viewed their role in assent
14as a progressive relationship between themselves, their parents, the
15researchers. Child participants expected that with increasing age they
16would have more control over their research participation. There was,
17however, varied opinion as to the correct age for children to assume
18consenting responsibilities for research. Many child respondents held the
19view that children might find it difficult to exercise their right to dissent to
20research consented by their parents. Third, the return of research results
21is a particularly important issue. Parents generally wanted to receive
22results, viewing them as a possible benefit of participation in a biobank.
23Some authors however understood this activity as having the capacity to
24infringe upon a child's autonomy. Finally, concern about risks, burdens and

1benefits of research participation was evident in the literature.

2Respondents in all studies wanted research to involve minimal risks and to
3not be burdensome. Parents supported biobank participation on condition
4that the associated benefits outweighed the risks.

5That consent and assent continue to be debated in paediatric research
6literature is unsurprising. While assent as defined by Alderson (2007)
7comprises a non-refusal or simple agreement without the understanding,
8discretion and legal validity associated with consent, consent invokes
9protection of one's integrity of body, mind and personal information (Miller
10and Boulton 2007). The latter is a concept with a dual ethical and legal
11nature (Brothers 2011). The consenting process in a paediatric research
12setting is ideally fashioned as a tripartite relationship between parents,
13children and researchers, though more probably involves value judgments
14by the parent or child's guardian rather than risk assessment or
15acceptance by the child. This is especially true where children are very
16young (e.g. in a birth cohort study) and parents give proxy consent based
17on a substituted judgment (i.e. the presumed judgment of child if he or
18she was competent) (Samuël, Knoppers et al. 2012). There is, of course,
19no guarantee that the guardian's decision is the same as the child's will.

20As children age it is necessary to examine their views on the consenting
21process to unlock pragmatic and ethical ways of handling this necessarily
22dynamic and changing relationship. Indeed, there are numerous examples
23in the field of paediatric biobanking of potentially good practice, though
24these are not often documented or verified by or with child participants
25themselves.

1 Understandable concerns about the potential risks of research
2 participation in this literature raise issues about definitions and
3 perceptions of risk. Framing potential dis-benefits of research as risks may
4 be itself problematic: absence of risk is not commensurate with the
5 absence of cost (Williams 2012). There may, for example, be relational or
6 power impacts or costs. In as much as dissent is available for children –
7 they are able to interpret social behaviour and develop social expectations
8 of themselves (Davis 1998) – such dissent may be difficult in practical and
9 emotional terms. A child's dissent may be treated as non-cooperation; and
10 could bear a cost in the relational dynamic within the tripartite
11 relationship, a relationship involving important power negotiations.
12 Arguably, inequalities of power will always exist for children involved in
13 research (Harden, Scott et al. 2000). While children are active social
14 agents, their lives are in many ways determined for them (Neale and
15 Smart 1998). The aim of including children in research and in debates
16 about research is to avoid the situation whereby 'children have a voice but
17 adults control the conversation' (Shanahan 2007). Irwin (2006) proposed
18 handling this power imbalance in research through the use of an
19 emancipatory model; one having the capacity to increase the children's
20 influence over research without necessarily suggesting that the children
21 should take over the research as is the case with participatory action
22 research (Alderson 2007). Emancipatory research within paediatric
23 biobanking addresses issues of power and respect and has the potential to
24 inform its practice by taking into account the children's' perspectives on
25 the research.

1The return of research results is keenly debated in the broader literature
2about biobanking. Guidelines for return of results routinely recommend
3that results should only be returned if they have analytic validity, clinical
4validity, and action-ability, and that the results themselves meet criteria
5related to severity of outcome (Hens, Van El et al. 2012, Knoppers, Zawati
6et al. 2012, Wolf, Crock et al. 2012). The return of results is sometimes
7viewed as a way of benefit sharing in research and by extension is an
8incentive for participation (Tabor, Brazg et al. 2011). In the papers
9reviewed here, parents in tissue banks expressed a belief that research
10participation lead to the development of new medications that could
11benefit them and their children. This perspective may propagate the
12expectation and misconception of therapeutic gain from research with a
13primarily scientific and non-therapeutic aim (Halverson and Ross 2012). In
14population biobanks, the equivalent misconception may be of the
15potential diagnostic benefits of research participation (Clayton and Ross
162006). Brothers (2011) argues that biobanks, especially those with de-
17identifiable resources, are not designed with the aim of, or capacity for,
18returning health-related results; also referred to as incidental findings.

19The studies in this review presented parental and adult perspectives that
20positioned children as lacking (if only in part) the capacity to make
21decisions about themselves and how they interact in the world.
22Historically, children have been viewed in contrast to adults, typically
23framed as victims or deviants when their views or performance differ from
24those of adults (Hood, Kelley et al. 1996). Paediatric research has been
25known to ascribe incompetence to children in a similar fashion, with the

1result that it is typically easier to prove a child's incompetence than it is
2for them to display their competence. And yet by the age of 5 years, a
3child has already developed a lifelong understanding of self, others,
4relationships and time (Alderson 2007, Uprichard 2008). Alderson (2007)
5argues children not only value interpersonal relationships, but they also
6have the ability to act responsibly and maintain these relationships.
7Children display sensitivity about differences in age and are generally
8eager to grow older due to their anticipation of a change in status (Bühler-
9Niederberger 2010). Ageing throughout childhood involves a relationship
10between body, self and society (Uprichard 2008). The creation and
11reproduction of youth sub-cultures selectively adopt and reject adult rules
12and interpretations (Shanahan 2007). Children appropriate information
13from the adult world and use it to constitute their own realities (Shanahan
142007). Although children borrow their parents' constructions (e.g. risk),
15they ultimately negotiate their own understandings (Hood, Kelley et al.
161996). Swartling et. al. (2011) have argued that the development of
17appropriate frameworks for research with children will only occur when
18adequate opportunities are available for adults to consult with children
19and consider as valid children's experiences and views. On this basis, the
20current literature on participant experience in paediatric biobanking is
21very much still in its infancy.

225. **Conclusion**

23In as much as the papers under review identified key themes in the views
24of stakeholders' participating in paediatric biobanks, very little of the
25evidence came from or could clearly be identified as coming from

1children. Moreover, much of the literature involving children and scientific
2and medical research is more generally contextualised within ethical
3debates (Harden, Scott et al. 2000) and assumes a largely protectionist
4perspective (Shanahan 2007). This social construction of children's
5vulnerability gives rise to the perception that children as research subjects
6are 'out-of-bounds', with the result that their voices are silenced (Moran-
7Ellis 2010). This situations propagates a dependency model and
8undermines the status of children as individuals in their own rights
9(Shanahan 2007). While there remains a need to balance inclusion of
10children in research with their protection (Avard, Silverstein et al. 2009), it
11is important to consider how efforts made to protect children may
12unintentionally protect the power of adults. But the role of children in
13research is evolving and the shift from their being silent and obedient
14subjects to autonomous and articulate participants (Alderson 2007)
15means appropriate biobanking practices involving children will occur as
16this stakeholder group is actively consulted (Goodenough, Williamson et
17al. 2004). Children have sophisticated understanding of concepts and
18issues surrounding genetics (Anderson, Stackhouse et al. 2011) and can
19contribute to complex policy debates on the topic (Grover 2004). Their
20virtual absence from empirical research at present is a critical omission to
21future development of paediatric biobanking policy.

22Limitations of the study

23This paper examined stakeholders' perceptions of paediatric biobanks by
24aggregating studies of general population biobanks involving mainly or

1only children with studies of disease-specific tissue banks of the same age
2group. Although this aggregation helped overcome the limited evidence
3base, our analysis was in turn unable to distinguish between possible
4differences in attitudes among stakeholders in biobanks and those among
5stakeholders in disease-specific tissues banks. Our findings suggested
6attitudes between the two groups converge towards similar themes,
7though this finding could be challenged as more studies emerge. Our
8analysis was also limited in that the majority of the studies included were
9based stakeholders’ hypothetical ideas rather than direct experience, and
10by the fact that the research designs of some studies suggested additional
11unpublished findings which could not be identified.

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1TABLE 1 SUMMARISING PAPERS REVIEWED

Authors	Type of Biobank	Methodology	Country	Key Findings
	<ul style="list-style-type: none"> actual vs hypothetical population biobank vs tissue bank Participants			
Brothers and Clayton (2012), (Brothers, Westbrook et al. 2013)	Hypothetical, Population biobank Parents (n=65)	Interviews	USA	Parents supported an opt-out model biobank in children and would allow their own child's sample to be included.
Brothers, Westbrook et al. (2013)	Actual , Population biobank Parents (n=237)	Interviews	USA	32.9% of participants were familiar with opt-out biorepository, while 92.4% approve of it based on a brief description.
Dixon-Woods, Wilson et al. (2008)	Actual, Tissue bank Children and parents (n=72)	Interviews	UK	Participants considered themselves to be members of a trusted community where values and interests were shared.
Goldenberg, Hull et al. (2009)	Hypothetical, Tissue bank Adults (n=1186)	Survey (telephone)	USA	67% would not be concerned about the use of childhood samples upon reaching adulthood. Concerned respondents were more likely to be more private about their medical records, less trusting of medical researchers, or African-American.
Goodenough, Williamson et al. (2003)	Actual, Population biobank Children (n=23)	Focus groups	UK	Children aged 8-10 years had valuable contributions to offer on their perceptions of participation in non-therapeutic longitudinal research.
Goodenough, Williamson et al. (2004)	Actual, Population biobank Children (n=40)	Interviews + focus groups	UK	Children have concerns over activities they are asked to take part in as research participants. Research participation fostered a feeling of 'being special' among children.
Halverson and Ross (2012), (Halverson and Ross 2012)	Hypothetical, Population biobank Parents (n=45)	Deliberative engagement + survey	USA	There was strong interest in receiving results, which was a main motivator for participation. The trust they had on the research would determine their enrolment.
Halverson and Ross (2012), (Halverson and Ross 2012)	Hypothetical, Population biobank Parents (n=45)	Deliberative engagement + survey	USA	Most participants stated they would enrol themselves and their children in a biobank. Some opposed enrolling children, particularly children unable to consent.

Harris, Ziniel et al. (2012)	Actual, Tissue bank Parents (n=19)	Focus groups	USA	Parents hoped to receive research results that would help them better understand their children's conditions or contribute to scientific knowledge.
Hens and Dierickx (2010), (Hens, Nys et al. 2010)	Hypothetical, Population biobank Teenagers and adults (n=76)	Focus groups	Belgium	There was a willingness to contribute tissue to research. Participants thought there was need for confidentiality protections. People expected to receive results that could be relevant to them.
Hens and Dierickx (2010)	Hypothetical, Population biobank Researchers (n=10)	Interviews	Belgium, UK, Saudi-Arabia	Long consent forms weren't preferred. Proper privacy and data protection was a need. Good communication considered important. Research on children needed to be for pediatric conditions.
(Hens and Dierickx 2010), Hens, Nys et al. (2010)	Hypothetical, Population Biobank Teenagers and adults (n=76)	Focus groups	Belgium	Research had to benefit and not burden children. Parents needed to engage their children in the decision-making. There was a need for re-contact upon maturity.
Hens, Snoeck et al. (2010)	Hypothetical, Population biobank Researchers (n=64)	Survey (Questionnaire)	Belgium	76.5% thought children should assent when they can comprehend; 51% estimated this to be aged 16-18 years.
Jackson, DIXON-WOODS et al. (2009)	Hypothetical, Tissue bank Researchers (n=331)	Survey (Questionnaires)	UK	100% were in favour of using tissue samples from children with cancer for research. 90% said both parent and child should consent. 94% supported 'generic' rather than 'specific' consent.
Kaufman, Geller et al. (2008)	Hypothetical, Population biobank Adults (n=141)	Focus groups	USA	Respondents were concerned with: minimizing children's fear, pain, and burdens; whether to include young children; and how to obtain children's assent.
Klima, Fitzgerald-Butt et al. (2013)	Actual, Tissue bank Children (n=378)	Survey (Consent assessment form)	USA	Parents understood consent for participation, purpose of study, and lack of direct benefit. Conversely, they least understood the indefinite storage of DNA, possible risks of participation, and that study was not for therapy.
Lemke, Halverson et al. (2012)	Hypothetical, Population biobank Parents (n=45)	Deliberative engagement + survey	USA	Focus group themes were: interest in biobank participation, broad consent and re-contact; trust in biobanking; and receiving research results. Survey data indicated same degree of interest in receiving results about themselves and their children.
McMurter, Parker et al. (2011)	Hypothetical, Tissue banking Parents (n=100)	Survey (Questionnaire)	Canada	89% agreed to have tissue sent anywhere for paediatric aims. 76% would permit genetic research even if no impact was anticipated. 41% would not allow painful, re-search procedures, while 15% would allow regardless of the child's dissent.

Neidich, Joseph et al. (2008)	Hypothetical. Population biobank Parents (n=239)	Survey (oral)	USA	Caucasians were the most willing to enroll their children into a pediatric biobank. Most respondents expressed optimism the results would yield significant benefits that would be distributed fairly.
Papaz, Safi et al. (2012)	Actual, Tissue bank Children and adults (n=3278)	Consent forms	Canada	Leading causes for refusal of consent were lack of interest in research 43%, feeling overwhelmed clinically 14%, and discomfort with genetics 11%.
Ries, LeGrandeur et al. (2010)	Actual, Population biobank Researchers (n=14)	Interviews	Canada, Denmark, England, France, Netherlands, USA	None adopted blanket consent for future use of samples/data. Ethics review of new studies a common requirement. Studies following children past early childhood sought assent/consent as the child matured.
Williamson, Goode-nough et al. (2004)	Actual, Paediatric biobank Children (n=167)	Interviews + focus groups	UK	Children's views are important in research and yet they underestimate the amount of control they have in it. Questioned parental rights to long-term use of children's samples.

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